



CASE REPORT

Isolated non-Hodgkin's lymphoma of the mandible

Derek M. Steinbacher *, Robert W. Dolan ¹

The Departments of Oral and Maxillofacial Surgery, Massachusetts General Hospital, Boston, Massachusetts and Otolaryngology, Lahey Clinic, Burlington, MA 02114, USA

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KEYWORDS

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Summary Primary non-Hodgkin's lymphoma (NHL) is rare. When it does occur, mandibular NHL typically manifests similar to an odontogenic process. This results in delayed diagnosis and treatment. We present a 53-year-old male who was erroneously treated for a dental process for several months before the correct diagnosis of diffuse large B-cell lymphoma was made. Fortunately, the solitary bony lymphoma had not disseminated and management by chemo-radiation allowed for disease eradication and new, post-treatment, bone formation. The purpose of this report is to describe a rare case of NHL of the mandible, explore the diagnosis and workup, and discuss treatment strategies.

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Introduction

Primary non-Hodgkin's lymphoma (NHL) is found outside of nodal tissues in 24–45% of cases.^{1,2} Common extranodal sites include the GI tract, skin, and, less often, bone.¹ In the head and neck, Waldeyer's ring, oral mucosa, salivary glands, paranasal sinuses, laryngeal tissue, and osseous structures have been found to exhibit primary NHL.³

The mandible accounts for only 0.6% of isolated malignant non-Hodgkin's lymphomas.⁴ When it does occur, mandibular NHL typically manifests similar to an odontogenic process. This results in a delay in diagnosis. The purpose of this report is to describe a rare case of NHL of the mandible, explore the diagnosis and workup, and discuss treatment strategies.

Case report

A 53-year-old male presented with a past history of vague pain along his right mandible. He underwent extraction of a mandibular molar tooth by an outside practitioner for a presumed odontogenic process. Following extraction, his symptoms stabilized and improved over the next three months. He then began noting pain in the vicinity of the previous extraction site and right lower lip numbness developed. This was thought to be "osteomyelitis" or an "abscess" and he was treated with a course of antibiotics. Subsequently, a fullness and fleshy mass was noted in the buccal sulcus in the same region and progressively enlarged. The area was biopsied and was found to be diffuse large B-cell lymphoma with tumor cells positive for CD20, CD79a, CD30, and Ki-1 (Fig. 1).

He was then referred for definitive management 10-months following his initial extraction. Workup included chest X-ray, CT scans (Fig. 2), PET scan (Fig. 3), bone scan,

* Corresponding author. Tel.: +1 617 523 3411.

E-mail address: dsteinbacher@partners.org (D.M. Steinbacher).

¹ Lahey Clinic, Department of Otolaryngology.

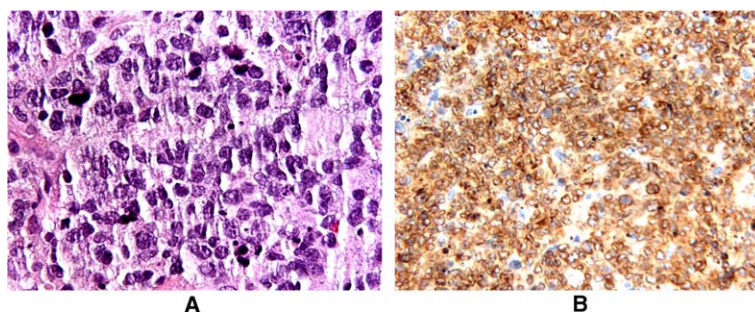


Figure 1 (A) Hematoxylin and eosin stain consistent with diffuse large B-cell lymphoma. (B) CD79A immunoperoxidase stain.

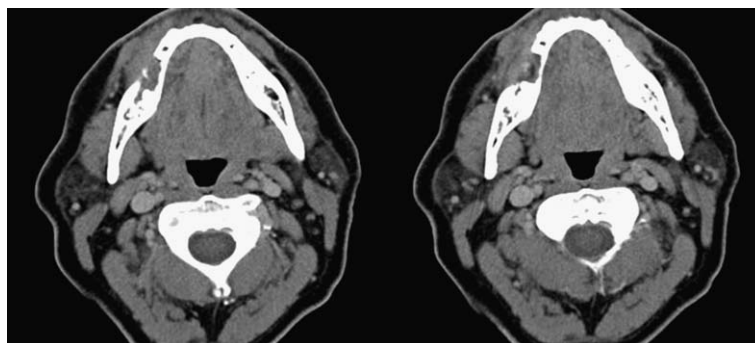


Figure 2 CT scan demonstrating the osteolytic focus in the right mandible with buccal cortical erosion.

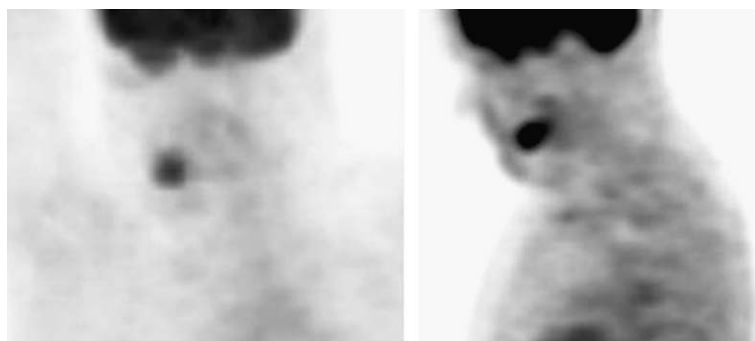


Figure 3 PET scan images showing the hyperintense right mandibular lesion.

and bone marrow aspirate. It appeared that his lymphoma was localized to the right mandible, without diffuse disease or nodal involvement. He therefore was categorized as stage IAE, primary mandibular NHL. It was the recommendation of oncologist that he undergo four cycles of CHOP Rituxan followed by radiotherapy. He attained a complete response and following therapy there appeared to be new healthy bone formation.

Discussion

NHL of bone is rare, representing only 5% of all extranodal lymphomas.⁵ The cell architecture and histology is indistinguishable from nodal or lymphoid derived lymphomas.⁶ Intrabony lymphomas were first described by Parker and Jackson.⁷ They arise from the medullary cavity and manifest as a localized, solitary lesion. To be classified as a primary NHL of bone there must be no evidence of visceral or lym-

phatic involvement and no distant metastases for at least 6 months following diagnosis.⁸ Primary mandibular involvement makes up about 5% of all NHL of bone and accounts for 8% all mandibular tumors.⁶

Malignant NHL of jaws affects adults in the 4th–5th decades with a male to female ratio of approximately 3:2, although some reports suggest a female predilection.^{5,6} The clinical presentation of mandibular NHL usually involves localized bone swelling, tooth resorption or mobility, mass in an extraction socket, pain, pathologic fracture, and often anesthesia or paresthesia along the distribution of the inferior alveolar nerve.^{3,4,6,9–14} Paresthesia along the inferior alveolar nerve distribution is common in NHL of the mandible with reports ranging from 20–100%.¹⁵

The diagnosis of NHL of the mandible is frequently delayed or erroneously diagnosed as dental disease because the clinical presentation mirrors that of an odontogenic process or localized osteomyelitis. There is an average elapse of 10-weeks between initial presentation and diagnosis.¹¹

There are no radiographic pathognomonic findings. Features are usually that of non-specific osteolysis. Panoramic films can show loss of cortical definition or widening of the mandibular canal, widening of the mental foramen, loss of lamina dura, or widening of periodontal ligament.¹⁶ Buccal or lingual cortical destruction is often found, but several reports note that NHL of the mandible can manifest as an extraosseous soft tissue mass with only minimal cortical destruction observable on plain films.¹⁶

Typically, the symptoms continue to persist in the face of attempted pharmacologic or dental therapies. This leads most practitioners to take tissue for sampling and pathologic diagnosis. Histopathologic evaluation, together with immunophenotypic and cytogenetic studies, elucidate the pattern of involvement and histologic type. Diffuse large B-cell lymphoma (formerly known as diffuse histiocytic lymphoma) is the most common subtype of NHL, including primary mandibular NHL.^{15,17}

When tissue diagnosis confirms NHL of the mandible, determination must be made regarding origination and spread of the tumor. The work up should assess the extent of disease and allow for accurate staging. Following clinical examination and palpation of nodes, computed tomography is essential to exclude visceral or nodal involvement. A skeletal survey is usually not warranted without the presence of bone pain or related complaints. PET scanning is reported to be comparable to CT and provides no additional information than the typical work-up.¹⁸ Laboratory studies are typically non-specific, although elevated lactate dehydrogenase is observed as a poor prognostic factor.¹⁹

Staging takes into account the involved site and degree of dissemination. Primary NHL of bone, as a single focus in an extranodal site, is categorized as stage IE. For all-comers, stage I NHL has a 5-year survival rate of 70% and the median survival time for IE is 10 years.⁹ The 5-year survival rate for stage IE NHL of the maxillo-mandibular region is reported to be approximately 50%.⁹ Survival declines to 20% and below for stages II–IV.

Treatment for primary lymphoma of the mandible typically consists of a combination of chemotherapy and radiotherapy. It is important to note that radiologic appearance does not always correlate with the true extent of the lesion. The radiation field should incorporate the ipsilateral mandible and regional lymph. Radiation alone may be curative in early lesions. Adjuvant chemotherapy is useful especially in histopathologic variants suggesting aggressive behavior. The role of surgery is for biopsy purposes and for the control of persistent or recurrent local disease.

We present a case of isolated mandibular NHL, stage IE. This is a rare condition but one that should be considered when patients have persistent dental symptoms recalcitrant to standard treatment. Fortunately, in this case, the solitary bony lymphoma had not disseminated and management by chemo-radiation allowed for disease eradication and new, post-treatment, bone formation.

References

- Freeman C, Berg JW, Cutler SJ. Occurrence and prognosis of extranodal lymphomas. *Cancer* 1972;**29**:252.
- Temmin L, Baker H, Amanguno H, Madda JP, Sinowatz F. Clinicopathological features of extranodal lymphomas: Kuwait experience. *Oncology* 2004;**67**(5–6):382–9.
- Ikeda T, Kanaya T, Matsuda A, Motohashi K, Tanaka H, Kohno N, et al. Clinicopathologic study of non-Hodgkin lymphoma in sinonasal and hard palate regions in 15 Japanese cases. *ORL J Otorhinolaryngol Relat Spec* 2005;**67**(1):23–9.
- Longo F, De Maria G, Esposito P, Califano L. Primary non-Hodgkin's lymphoma of the mandible: report of a case. *Int J Oral Maxillofac Surg* 2004;**33**:801–3.
- Toliusis V, Pamerneckas A, Petrulis A, Tamulaitis G, Pilipavicius A, Pijadin A. Primary non-Hodgkin's lymphoma of bone. *Medicina (Kaunas)* 2004;**40**(4):379–85.
- Huvos AG. *Bone tumours. Diagnosis, treatment, and prognosis*. 2nd ed. Philadelphia, London, Toronto: WB Saunders & Co.; 1991., pp. 625–37.
- Parker F, Jackson H. Primary reticulum cell sarcoma of bone. *Surg Gynecol Obstet* 1939;**68**:45–53.
- Piatelli A, Croce A, Tete S, Artese L. Primary non-Hodgkin's lymphoma of the mandible: a case report. *J Oral Maxillofac Surg* 1997;**55**:1162–6.
- Bachaud JM, Cappin D, Douchez J, et al. Primary malignant lymphoma of the mandible: a study of 3 cases and review of the literature. *Rev Stomatol Chir Maxillofac* 1992;**93**:372.
- Barber DH, Stewart JCB, Baxter WD. Non-Hodgkins lymphoma involving the inferior alveolar canal and mental foramen: report of a case. *J Oral Maxillofac Surg* 1992;**50**:1334.
- Parrington SJ, Punnia-Moorthy A. Primary non-Hodgkin's lymphoma of the mandible following tooth extraction. *Br Dent J* 1999;**187**:468–70.
- Liu RS, Liu HC, Bu JQ, et al. Burkitt's lymphoma presenting with jaw lesions. *J Periodontol* 2000;**71**:646.
- Park YW. Non-Hodgkin's lymphoma of the anterior maxillary gingival. *Otolaryngol Head Neck Surg* 1998;**119**:146.
- Nocini P, Muzio LL, Fior A, et al. Primary non-Hodgkin's lymphoma of the jaws: immunohistochemical and genetic review of 10 cases. *J Oral Maxillofac Surg* 2000;**58**:636.
- Pazoki A, Jansisyananont P, Ord R. Primary non-Hodgkin's lymphoma of the Jaws: report of four cases and review of the literature. *J Oral Maxillofac Surg* 2003;**61**:112–7.
- Yamada T, Kitagawa Y, Ogasawara T, et al. Enlargement of the mandibular canal without hypesthesia caused by an extranodal non-Hodgkin's lymphoma: a case report. *Oral Surg Oral Med Oral Path* 2000;**89**:388.
- Armitage J, Weisenburger D. New approach to classifying Non-Hodgkin's lymphomas: clinical features of the major histologic subtypes. *J Clin Oncol* 1998;**16**:2780.
- Kostakoglu L, Leonard JP, Kuji I, et al. Comparison of fluorine-18 fluorodeoxyglucose positron emission tomography and Ga-67 scintigraphy in evaluation of lymphoma. *Cancer* 2002;**94**:879.
- Dumontet C, Drai J, Bienvenu J, et al. Profiles and prognostic values of LDH isoenzymes in patients with non-Hodgkin's lymphoma. *Leukemia* 1999;**13**:811.